

## CASE REPORT

# Gallstone ileus, a rare cause of intestinal occlusion. A case report

## Íleo biliar recurrente, una causa infrecuente de oclusión intestinal. Presentación de un caso clínico

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### Abstract

Recurrent gallstone ileus has a recurrence of 2-8.2% with a mortality of 12-20%, secondary to an enteric or cholecystic gallstone. A male patient with a diagnosis of intestinal occlusion secondary to biliary ileus and cholecystoduodenal fistula, performing enterotomy and closure in two planes with drainage placement. Two months after presenting the clinical of intestinal occlusion, medical management began and an abdominal tomography was performed, finding an image suggestive of recurrent gallstone ileus, treated with laparotomy.

**Keywords:** Intestinal occlusion. Gallstone ileus. Cholecystoduodenal fistula.

### Resumen

El íleo biliar recurrente tiene una frecuencia del 2-8.2% y una mortalidad del 12-20%, que se presenta de forma secundaria a un cálculo biliar entérico o colecístico. Varón que cursa con diagnóstico de oclusión intestinal secundaria a íleo biliar y fístula colecistoduodenal. Se realiza enterotomía y cierre en dos planos con colocación de drenaje. Dos meses después, el paciente presenta un cuadro clínico de oclusión intestinal, por lo que se inicia manejo médico y se realiza la correspondiente tomografía computarizada abdominal, encontrando una imagen sugestiva de íleo biliar recurrente, con manejo por laparotomía.

**Palabras clave:** Oclusión intestinal. Íleo biliar. Fístula colecistoduodenal.

### Introduction

Biliary ileus is a rare complication of biliary disease, accounting for 0.5% to 4% of all mechanical small bowel obstructions. It is due to repeated episodes of cholecystitis through an internal fistula between the gallbladder and the duodenum, stomach, or colon, being the most common cause cholecystoduodenal fistula<sup>1</sup>.

The early diagnosis of biliary ileus can be achieved by combining clinical symptoms (epigastric pain with nausea and vomiting) with abdominal computed tomography findings revealing the presence of Rigler's triad (ectopic calculi, intestinal obstruction, and pneumobilia)<sup>2</sup>. Surgical management is the definitive treatment, which can be performed through enterolithotomy, along with cholecystectomy and fistula repair (1-stage surgery), or through simple enterolithotomy (the most

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widely performed surgical procedure). The medical literature currently available supports enterolithotomy alone as a definitive therapy to treat biliary ileus, as it is reported that retained calculi after initial treatment with enterolithotomy alone will remain asymptomatic in 80% to 90% of the patients<sup>3</sup>, and it has a low mortality rate (12%) compared to cholecystectomy and fistula repair (17%). However, there is a risk of recurrence involved<sup>4</sup>.

Overall, the rate of recurrence of biliary ileus ranges from 2% to 8.2%, and mortality rate from 12% to 20%<sup>5</sup>. Recurrence is often due to an unidentified enteric or cholecystic gallstone at the time of the initial surgery<sup>6</sup>. Nearly 50% of all recurrences occur within the first month after the initial surgery, and the remaining 50% within the first 2 years, with reports of up to 9 years<sup>7</sup>.

Recurrent gallstone most commonly occurs in the ileum (49.5%), but it has also been reported in the colon and rectum in up to 3.8% of the cases<sup>8</sup>. The mean size of the obstructing stone is 3.6 cm (ranging from 1 cm to 5-6 cm). The smallest stone ever reported causing obstruction in the small intestine was 1.5 cm, and 3 cm in the large intestine. The largest stone ever reported in the medical literature currently available had a diameter of 17.7 cm. Nonetheless, the size of the stone does not seem to be associated with the obstruction site<sup>9</sup>.

At the time of the second laparotomy due to recurrent biliary ileus, intestinal obstruction resulted from a single stone in 89.2% of the cases. Two or more stones were reported in 10.7% of the cases. Similarly, in 42.5% of the cases, the site of impaction was distal to the previous enterolithotomy. On the other hand, in 22.6% of the cases, the stone impacted at the previous repair site, while in 34.6% of the cases, the impaction site was proximal to the site of previous enterolithotomy<sup>10</sup>.

Recurrent biliary ileus should be considered in any patient with a past medical history of biliary ileus experiencing a second episode of small bowel obstruction<sup>11</sup>. Despite its size, the abdominal physical examination cannot identify non-obstructing stones in a significant number of the cases<sup>12</sup>. The persistence or recurrence of symptomatic biliary tract disease after surgery should lead to consideration of a second procedure for fistula repair and cholecystectomy<sup>13</sup>.

Overall, the basis for preventing recurrence is to make meticulous efforts before and after surgery to identify and remove stones  $\geq 2$  cm, such as performing perioperative ultrasounds to detect residual stones



**Figure 1.** Abdominal x-ray at admission showing multiple air-fluid levels suggestive of intestinal obstruction.

inside the gallbladder, and palpating all portions of the intestine and the gallbladder during surgery to make sure that there are no retained stones left, which typically prevents the problem. The presence of cylindrical or faceted stones during surgery should alert the surgeon on the possibility of new stones<sup>14</sup>.

## Case report

This is the case of a 42-year-old man with a history of appendectomy reported in 2008 and exploratory laparotomy in May 2014 due to intestinal obstruction, with adhesion release and appendectomy.

The patient presented to the ER reporting the onset of his current condition 3 days prior to admission, with epigastric pain of intensity 6/10, radiating to the right hypochondrium, along with nausea, multiple episodes of vomiting with gastric content, and oral intolerance. He denied passing gas and having bowel movements. Upon admission, his blood pressure level was 100/55 mmHg; heart rate, 54 bpm; respiratory rate, 19 rpm; and temperature, 36°C. The physical examination revealed the presence of a distended abdomen due to adipose tissue and intestinal gas, which was soft, depressible, diffusely tender upon palpation, and without any signs of peritoneal irritation. Lab test results: leukocytes, 18 600; neutrophils, 83.3%; lymphocytes, 8.9%; hemoglobin, 18.7 g/dL; hematocrit, 60.60%; platelets, 437 000; prothrombin time, 13.2 s; partial thromboplastin time, 36.6 s; INR (International Normalized Ratio),

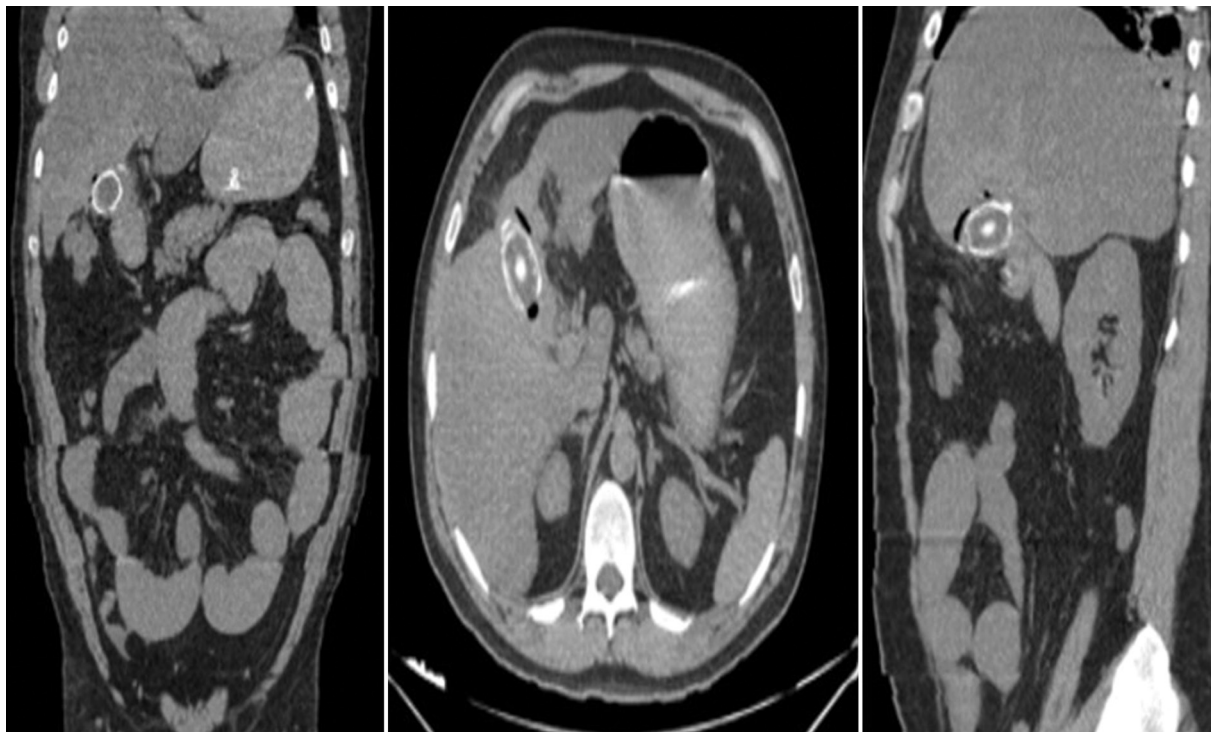


**Figure 2.** Abdominal computed tomography 24 hours after admission. The gallbladder with calcified walls can be seen here, with an apparent gallbladder-duodenum fistula.

1.1; activity (percentage of activity), 77.6%; glucose, 206.4 mg/dL; blood urea nitrogen, 34.5 mg/dL; urea, 73.83 mg/dL; creatinine, 4.2 mg/dL; sodium, 133.5 mEq/L; potassium, 4.01 mEq/L; chloride, 91.3 mEq/L; calcium, 12.2 mg/dL; phosphorus, 2.2 mg/dL; total bilirubin, 0.8 mg/dL; direct bilirubin, 0.2 mg/dL; indirect bilirubin, 0.6 mg/dL; glutamic-oxaloacetic transaminase, 22 IU/L; glutamic-pyruvic transaminase, 33 IU/L; albumin, 6.3 g/dL; gamma-glutamyl transpeptidase, 104 IU/L; alkaline phosphatase, 139 IU/L; and lactate dehydrogenase, 195 IU/L. The abdominal x-ray performed at admission showed multiple air-fluid levels (Fig. 1).

Initial medical management included nasogastric tube placement, fluid control, IV antibiotic therapy with metronidazole 500 mg/8 h and ceftriaxone 1 g/12 h, antiemetics, and analgesics. The patient was re-evaluated 8 hours later, during which he experienced 3 episodes of diarrhea but still no passing of gas. He also showed a tendency towards tachycardia, and the nasogastric tube output consisted of 200 cc of intestinal content. However, he continued to have elevated nasogastric tube outputs of 1400 cc plus 1500 cc of intestinal content vomiting. An abdominal computed tomography was performed at 24

hours that revealed the presence of a gallbladder with calcified walls with solution of continuity with apparent connection to the duodenum, along with free perivesicular air. Intestinal loops showed adequate contrast passage to the rectal ampulla and limited intraluminal gas in the small bowel. Fat density changes in the small bowel mesentery were also seen, with a decrease of up to 50 HU compared to the remaining intraabdominal fat (Fig. 2).

Given the patient's current condition and the tomographic findings suggestive of choledocoduodenal fistula, biliary ileus was suspected. The exploratory laparotomy performed under general anesthesia revealed the presence of multiple firm and lax adhesions between the omentum and the intestine, with nearly 100 cc of reactive fluid. A 3 cm × 3 cm gallstone was found 80 cm away from the ileocecal valve that was responsible for the small bowel distension (Fig. 3). Consequently, small bowel adhesiolysis followed by enterolithotomy were performed with subsequent primary closure in 2 planes in a surgical time of 1 hour and 40 min (Figs. 4 and 5).

Within the first 24 hours after surgery, the patient showed clinical symptom improvement, with the passage of gas and bowel movements. Additionally, he received partial



**Figure 3.** Intestinal obstruction site 80 cm away from the ileocecal valve.



**Figure 4.** Enterolithotomy and gallstone removal.

parenteral nutrition, pain management, and antibiotics. The intestinal transit contrast study performed 7 days after surgery confirmed passage towards the colon and sigmoid colon, which is why it was decided to start a liquid diet, with adequate tolerance, and progress gradually. The nasogastric tube was removed, and the patient was discharged uneventfully 10 days after surgery.

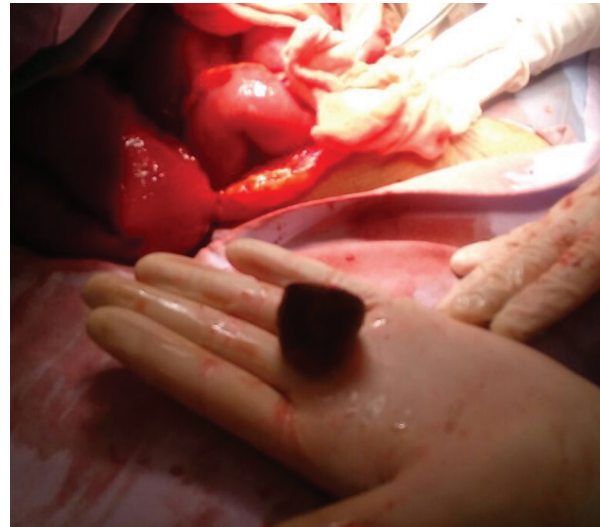


**Figure 5.** 3 cm x 3 cm gallstone, cause for the intestinal obstruction.

However, 10 weeks after the index biliary ileus event, the patient returned to the ER with symptoms that began 2 days prior to admission. He presented with abdominal colicky pain of mild intensity initially, which escalated to incapacitating pain with an intensity of 9/10. He experienced nausea, multiple episodes of vomiting with bilious content, no passage of gas in 24 hours, and a semiliquid bowel movement also 24 hours before admission. Additionally, he showed decreased appetite, weakness, fatigue, and overall poor general condition. His blood pressure was 129/90 mmHg; heart rate, 140 bpm; respiratory rate, 20 rpm, and temperature, 37 °C. On abdominal examination, the abdomen appeared distended due to intestinal gas, reduced peristalsis, diffuse tenderness upon palpation with voluntary and involuntary muscle resistance, and tympanic percussion in the upper abdomen, with no signs of peritoneal irritation. The lab test results at admission were: leukocytes 13 400; neutrophils, 73.7%; lymphocytes, 17.2%; hemoglobin, 13.20 g/dL; hematocrit, 42.10%; platelets, 340 000; prothrombin time, 13.6 s; partial thromboplastin time, 29.3 s; INR, 1.13; activity (percentage of activity), 29.3%; glucose, 101.6 mg/d; blood urea nitrogen, 15.40 mg/dL; urea, 32.96 mg/dL; creatinine, 0.7 mg/dL; sodium, 136.1 mEq/L; potassium, 3.49 mEq/L; chloride, 103.2 mEq/L; calcium, 9.2 mg/dL; phosphorus, 1.8 mg/dL; total bilirubin, 0.5 mg/dL; direct bilirubin, 0.2 mg/dL; indirect bilirubin, 0.3 mg/dL; glutamic-oxaloacetic transaminase, 14 IU/L; glutamic-pyruvic transaminase, 14 IU/L; albumin, 4.1 g/dL; gamma-glutamyl transpeptidase, 41 IU/L; alkaline phosphatase, 41 IU/L;

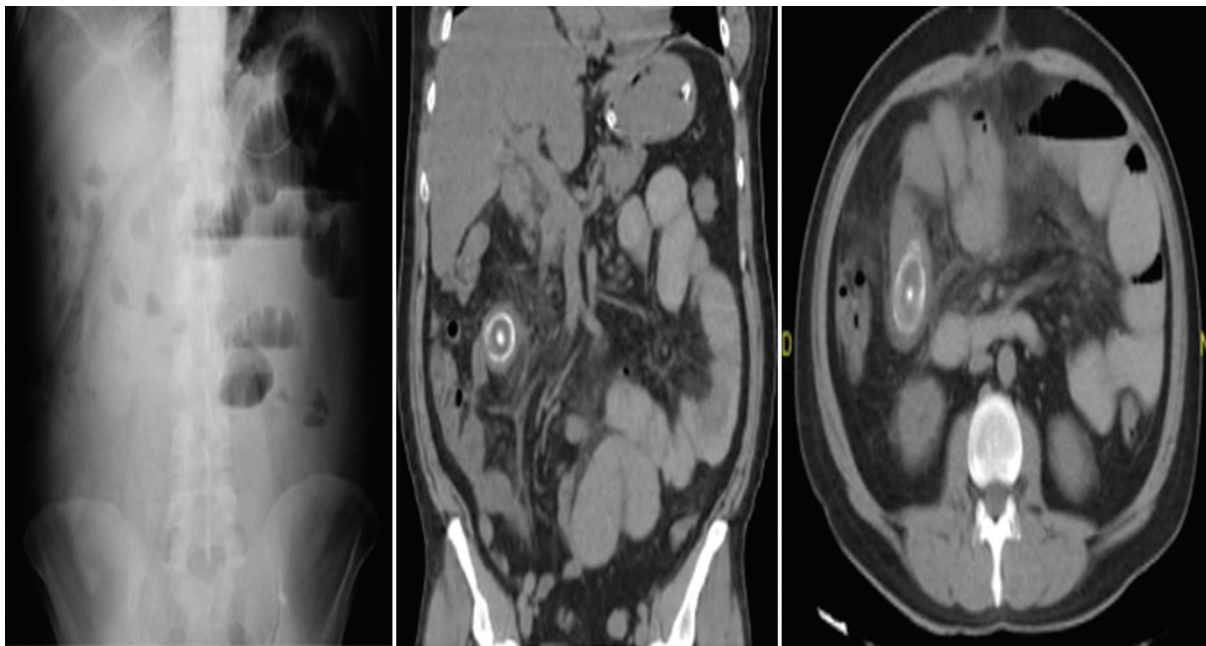


**Figure 6.** Abdominal x-ray showing signs of intestinal obstruction and abdominal computed tomography suggestive of a gallstone in the small bowel.

and lactate dehydrogenase, 124 IU/L. Similarly, the x-ray and the abdominal computed tomography performed (Fig. 6) showed signs of intestinal obstruction, with the CT scan revealing an image suggestive of a stone within the intestinal lumen.

Medical management was initiated, including fasting, placement of a nasogastric tube, and fluid control. The nasogastric tube output consisted of 50 cc of gastric content over 8 hours, which prompted an emergency surgical intervention request. The exploratory laparotomy performed revealed the presence of multiple firm adhesions between bowel loops and a single 8 cm × 9 cm stone inside the small bowel—although the exact location was not identified—completely occluded the intestinal lumen. Adhesion release, intestine resection at the obstruction site, and the creation of an ileostomy were performed during surgery, which went on for 4 hours and 50 min.

The patient had a favorable early postoperative course, with ileostomy outputs > 800 cc in 24 hours. Therefore, the patient was transitioned to a soft diet, which was well-tolerated, and with good postoperative disease progression. He remained on antibiotic therapy, and was discharged from the hospital 5 days after surgery without complications.

The patient had a satisfactory postoperative course at the follow-up performed outpatiently. Therefore, it was decided to schedule a procedure to restore

intestinal transit 4 months after the last laparotomy. A mechanical end-to-end entero-enteroanastomosis was performed 160 cm away from the angle of Treitz and 50 cm away from the ileocecal valve. The patient remained on a fasting regimen, on antibiotics, IV fluids, glucose, and electrolytes, with a favorable recovery. He began to tolerate oral intake, passed gas through the rectum, and experienced bowel movements 72 hours after surgery. The patient had a favorable postoperative course and was uneventfully discharged from the hospital with outpatient follow-up.

## Discussion

Overall, the rate of recurrence of gallstone ileus ranges from 2% to 8.2%<sup>15</sup>, and mortality rate between 12% and 20%<sup>16</sup>. It often occurs following an episode of acute cholecystitis, especially in elderly patients and women who present with acute abdominal symptoms<sup>7</sup>.

Ours was the case of a male patient who experienced recurrent gallstone ileus 10 weeks after the initial operation that revealed the presence of a second stone. It has been previously reported that recurrent gallstone ileus often occurs within 6 months after the initial operation, with the fastest recurrence recorded

happening within the first 30 days<sup>10</sup>. Very few cases of earlier recurrences have been reported in the medical literature currently available.

During the laparotomy exploration, multiple intestinal adhesions with difficult separation and unclear exposure of the gallbladder were revealed. Therefore, delayed cholecystectomy within 2 to 6 months is still under discussion<sup>7</sup>. In some favorable cases, if cholecystectomy seems straightforward, enterotomy for stone removal can be performed while the perioperative radiological follow-up of the main biliary tract is being conducted. Also, transverse suture can be used in the fistula located at the upper edge of the first section of the duodenum in 1 or 2 planes after refreshing its edges and with contact drainage. Whether or not to proceed with cholecystectomy in the presence of a cholecystoduodenal fistula is highly controversial in the current medical literature available. However, although there is substantial data to avoid this measure due to the high morbidity rate reported, it is not considered a contraindication *per se*<sup>2</sup>.

A thorough examination is highly recommended during the initial laparotomy to prevent recurrent gallstones. However, in this case, although the intestines were examined during the initial surgery to detect additional stones, the second stone was likely located in the gallbladder or the duodenum, making it difficult to find.

## Conclusions

Recurrent gallstone ileus is a rare complication of biliary pathology associated with a high morbidity rate when it occurs and should be considered in any patient with a past medical history of gallstone ileus with a second episode of small bowel obstruction. The cause of recurrent ileus may be due to an unidentified enteric or cholecystic gallstone at the time of the initial operation. Therefore, a thorough examination of all intestinal segments and the gallbladder during surgery is advised, along with perioperative ultrasound to detect the presence of any residual stones inside the gallbladder. These measures guarantee the patient's own safety in the presence of recurrences.

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## Conflicts of interest

None declared.

## Ethical disclosures

**Protection of human and animal subjects.** The authors declare that no experiments were performed on humans or animals for this study.

**Confidentiality of data.** The authors declare that they have followed the protocols of their work center on the publication of patient data.

**Right to privacy and informed consent.** The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

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